Case Report

A primary meningioma of the lumbar spine with neck metastasis

Chih-Wei Sung[1], Kevin Li-Chun Hsieh[2,3], Yi-Jie Kuo[4,5]

[1]Department of Emergency Medicine, National Taiwan University Hospital Hsin-Chu Branch, Hsinchu, Taiwan, [2]Department of Medical Imaging, Taipei Medical University Hospital, Taipei, Taiwan, [3]Department of Radiology, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan, [4]Department of Orthopedic Surgery, Wan Fang Hospital, Taipei Medical University, Taipei, Taiwan, [5]Department of Orthopedic Surgery, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

Context: Approximately 25% of all primary spinal cord tumors are meningiomas, and 80% of these tumors occur in the thoracic region. Few meningiomas of the lumbar spine have been presented. Extracranial metastasis of meningioma occurs extremely rare, only in about 0.1% of meningiomas. Even metastasis, the sites are seldom seen in deep soft tissue. We reported a woman original meningioma in the lumbar spine with distal deep neck metastasis.

Findings: A 59-year-old patient suffered from severe right drop foot, numbness, and radicular pain for the previous 6 months. Computed tomography (CT) disclosed a huge, macrolobulated retroperitoneal soft-tissue lesion with a size of $14.9 \times 10.8 \times 17.7$ cm. Magnetic resonance imaging (MRI) further revealed a solid spinal intracanal tumor with moderate enhancement involving the right paraspinal region at the L2∼L5 level and the right iliac fossa. A meningioma was diagnosed with histological proof. Four months later, another metastatic meningioma in her left neck. She was managed conservatively without neurologic dysfunction.

Conclusions: To the best of our knowledge, such a large meningioma of the lumbar spine has not previously been reported in the literature. In this study, we demonstrated a rare spinal meningioma located in the lumbar spine primarily with secondary soft tissue metastasis.

Keywords: Meningioma, Spinal meningioma, Lumbar spine, Metastatic, Neck metastasis

Introduction

Meningiomas, dural-based tumors, arise from arachnoid cap cells and can be located in the intracranial or extracranial space wherever the dura is present. Most meningiomas are slow-growing low-grade tumors, regardless of whether they are pathologically malignant or not. Meningiomas of the spine are reported to account for 10% of all meningiomas and occur anywhere along the spinal cord, with approximately 15% of them in the cervical spine, 81% in the thoracic spine, and 4% in the lumbar spine.[1] Generally, meningiomas of the lumbar spine are seldom seen. In previous decades, few reports mentioned patients with large, typical meningiomas of the lumbar spine. We herein report a rare case of a large spinal meningioma in the L2∼5 segments in a middle-aged woman.

Correspondence to: Yi-Jie Kuo, Department of Orthopedic Surgery, Wan Fang Hospital, Taipei Medical University, 111 Xinglong Rd., Sec. 3, Taipei 11696, Taiwan. Email: benkuo5@tmu.edu.tw

Informed consent for the publication of this case including the photographs was obtained from the patient.

Case

A 58-year-old woman presented to the emergency room with right drop foot, numbness, and radicular pain for the previous 6 months. She denied incontinence or a history of a herniated intervertebral disc, but she had a medical history of hypertension, diabetes, coronary artery disease, and a pulmonary embolism. Her vital signs were stable at triage. On physical and neurological examination, the sensation of the right leg was intact, while the entire right low extremity was paralyzed. At the time of initial presentation, Computed tomography (CT) disclosed a huge, multi-lobulated retroperitoneal soft-tissue lesion ($14.9 \times 10.8 \times 17.7$ cm), involving the right L3∼L5 transverse foramina, L3 and L4 transverse processes, iliac fossa, and psoas and iliac muscles as

DOI 10.1080/10790268.2018.1564993

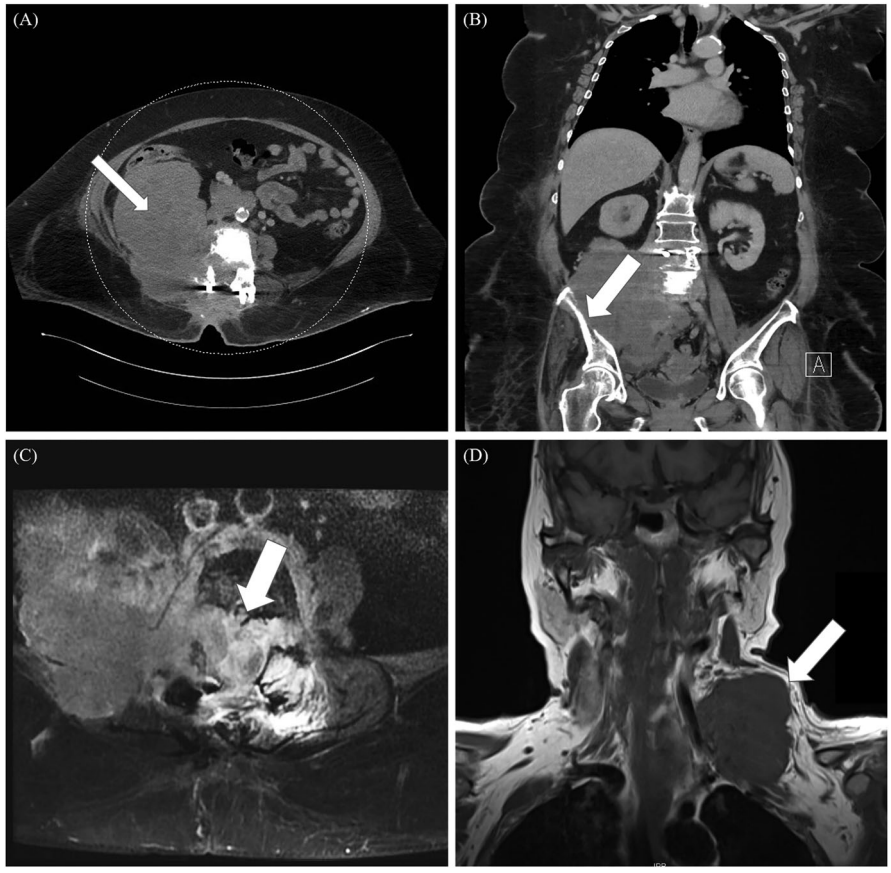

Figure 1 Contrast-enhanced computer tomographic (CT) (panel A: transverse view; panel B: coronal view) and contrast-enhanced T1-weighted magnetic resonance (MR) scans of the patient (panel C: axial view of lumbar spine, showing occupied the right iliac fossa by the solid intra-canal tumor; panel D: coronal view of neck).

shown in Fig. 1 (Panel A: transverse view; panel B: coronal view). In panels C, the contrast-enhanced T1-weighted magnetic resonance (MR) images further disclosed this solid intracanal tumor extending from the spinal canal to the right paraspinal region at the L2∼L5 level, which occupied the right iliac fossa (Panel C: axial view). Immunohistochemically, tumor cells were positive for vimentin and focally positive for the epithelial membrane antigen, and had a 5% proliferation index of Ki-67. A radiation oncologist was consulted, and maximal surgical debulking with adjuvant radiotherapy was suggested. However, the patient could not tolerate the entire treatment because the size was too large and widely extended into the abdominal cavity, multiple levels of the lumbar spine, and partial spinal roots. The high recurrence rate decreased the possibility of successful radical resection. Also, the patient had other comorbidities so that she

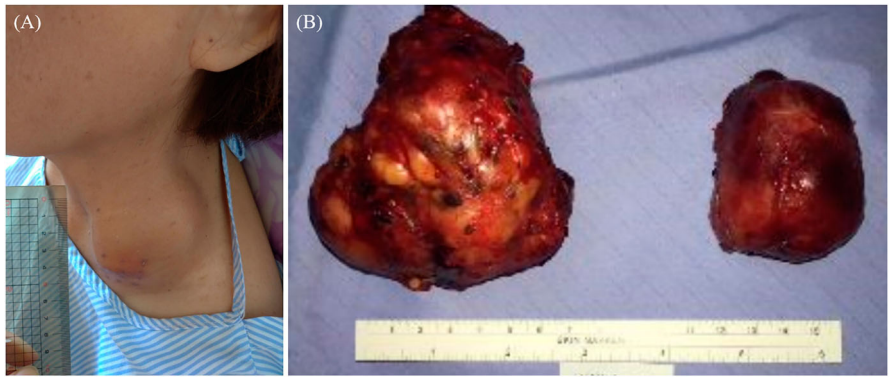

Figure 2 A neck mass which was proven metastatic meningioma (panel A: in appearance; panel B: tumor mass).

was at high risk with general anesthesia in surgical intervention. She was finally treated conservatively and radiotherapy was performed for 3 weeks in total. Additionally, a slow-release plus short-term morphine combination was administered for better pain control. She was finally discharged after almost 2 months of treatment. Four months later, she suffered from a neck mass about 4 × 6 cm (Fig. 2, panel A) in appearance. Excisional biopsy was performed and it pathologically revealed metastatic meningioma with atypical feature such as increased mitotic figures, tumor necrosis, and focal rhabdoid appearance (Fig. 2, panel B). A diagnosis of intra-dural extra-cranial meningioma with neck metastasis was finally made.

Discussion

Meningiomas, usually benign, originate from arachnoid cells and are mostly located in the intracranial compartment. Spinal meningiomas, which are characteristically intradural and extramedullary, are relatively rare and account for less than 10% of all meningiomas and 25% of all spinal cord tumors.[2] The incidence of intradural spinal tumors varies but is roughly 64 per 100,000 person-years, and these tumors account for 3% of primary central nervous system (CNS) tumors.[3] Women aged 40∼70 years are more vulnerable to spinal meningiomas than are men of any age. The peak age of occurrence for spinal meningiomas is 40∼70 years.[4] Spinal meningiomas can grow or metastasize to anywhere along the spinal cord where dura exists. Previous reports indicated that the thoracic spine (67%∼84%) is the most common location, which is more common than the cervical (14%∼27%) and lumbar spine (2%∼14%). Cohen-Gadol *et al*. reported their 21-year experience and showed that patients ≤50 years have a 40% incidence of spinal meningiomas in the cervical spine, especially in the upper cervical spine.[5] Levy and colleagues further reported that a preponderance of thoracic spine meningiomas occurred in female patients. Additionally, the location in relation to the spinal cord (anterior [15%], posterior [18%], or lateral [68%]) was clinically used to evaluate spinal meningiomas.[6] Chen *et al*. reported a lumbar clear cell meningioma with foraminal extension in a renal transplant recipient.[7] In 2018, Cunha *et al*. presented a case whose presentation was similar to a lumbar spinal meningioma but was eventually diagnosed as tophaceous gout.[8] The case we present is the first typical lumbar meningioma in recent years.

Clinically, manifestations in patients with a meningioma of the lumbar spine depend on the tumor location, in relation to the spinal cord transection and nerve roots. Neurological symptoms include back or radicular pain, motor weakness, sensory disturbances, and urinary or fecal incontinence as a late finding.[9,10] The diagnostic time varies person to person at between 1 and 2 years.[11] Our patient presented with radicular pain, numbness, and/or radiculopathy within just 6 months, probably due to the rapid progression of her large meningioma which also involved spinal roots of L2∼L5. For a diagnosis, plain film is usually normal except for bone erosion or calcification. MRI (gadolinium enhanced T1-weighted sequences and T2 sequences) is currently the standard imaging modality for spinal cord tumors, because it provides markedly excellent sensitivity and specificity, especially for spinal cord meningiomas. CT, however, plays an accessory role for its high resolution of bone and other solid organs in the abdomen. On MRI, characteristics of spinal meningiomas include well-circumscribed, broad-based dural attachment and/or a dural tail sign, which are similar signal characteristics to typical intracranial meningiomas. Spinal meningiomas typically demonstrate isointensity to slightly hypointensity on T1-weighted images, isointensity to slightly hyperintensity on T2-weighted images, and moderate homogeneous enhancement on T1-weighted images with gadolinium enhancement. Densely calcified meningiomas are sometimes hypointense on T1 and T2 images, and show only minimal contrast enhancement.[12] On CT, an isodense or moderately hyperdense mass may be revealed. Hyperostosis may also occasionally be seen. The CT presentation in our patient included a large mass with involvement of the right L3∼L5 transverse foramen, right pelvic region extension to the right L3 and L4 transverse processes, and right iliac wing invasion. No obvious calcification was found in the mass. MRI further revealed its huge size of 14.9 × 10.8 × 17.7 cm, hypointense necrotic part, and enhancement after gadolinium injection on a T1-weighted image.

These radiological characteristics are usually nonspecific. It is difficult for physicians to differentiate benign or malignant tumors simply on images. The "dural tail sign", for example, can be seen in intradural-extramedullary meningiomas but is also found in metastatic tumors and lymphomas.[13] A definite diagnosis largely depends on the pathology. Epithelial membrane antigen (EMA) is the most reliable immuno-marker of meningiomas, whereas some malignant tumors, such as hemangiopericytomas (HPCs), are negative for EMA.[14] In the current case, EMA was partially

positive, which was also compatible with a lumbar meningioma.

Surgical interventions may be definitive treatments for spinal meningiomas.[4] Even with a low complication rate of surgery of approximately 3%, individual outcomes depend on the size and location of the tumor and the preoperative neurologic state of the patient. Adjuvant radiotherapy and palliative radiotherapy have been suggested as being useful in cases of recurrent spinal meningiomas.[4,15] The most common risks are postoperative neurologic sequelae like motor weakness, sensory loss, and autonomic dysfunction. Overall, the recurrence rate for spinal meningiomas is low and ranges 0%∼10%.[16]

Extracranial metastasis of primary spinal meningioma occurs extremely rare, only in about 0.1% of the meningiomas.[17] Metastatic sites include the lung, pleura, and mediastinum. Few case reports documented deep soft tissue as a site of metastasis.[18] The mechanism of the metastasis remains unclear that soft tissue alone or other contiguous structures like lymph nodes and bone, is involved in metastasis.

In conclusion, primary lumbar meningiomas with neck metastasis are extremely rare. Although spinal meningiomas account for 1.2% of all meningiomas, almost all of them are located on the cervical or thoracic spine. Lumbar meningioma with neck metastasis is extremely rate.

Disclaimer statements

Contributors None

Funding None

Conflicts of interest Authors have no conflicts of interest to declare.

References

1 Mechtler LL, Nandigam K. Spinal cord tumors: new views and future directions. Neurol Clin. 2013;31:241–68.
2 Ley Valle A, Gando L, Codina A, Acarin PN. [Extradural spinal cord meningioma. Report of a case (author's transl)]. Rev Esp Otoneurooftalmol Neurocir. 1978;36:61–4.
3 Ostrom QT, Gittleman H, Liao P, Rouse C, Chen Y, Dowling J, Wolinsky Y, *et al*. CBTRUS statistical report: primary brain and central nervous system tumors diagnosed in the United States in 2007–2011. Neuro Oncol. 2014;16(Suppl 4):iv1–63.
4 Ravindra VM, Schmidt MH. Management of spinal meningiomas. Neurosurg Clin N Am. 2016;27:195–205.
5 Cohen-Gadol AA, Zikel OM, Koch CA, Scheithauer BW, Krauss WE. Spinal meningiomas in patients younger than 50 years of age: a 21-year experience. J Neurosurg. 2003;98:258–63.
6 Levy Jr. WJ, Bay J, Dohn D. Spinal cord meningioma. J Neurosurg. 1982;57:804–12.
7 Chen MH, Chen SJ, Lin SM, Chen MH. A lumbar clear cell meningioma with foraminal extension in a renal transplant recipient. J Clin Neurosci. 2004;11:665–7.
8 Ribeiro da Cunha P, Peliz AJ, Barbosa M. Tophaceous gout of the lumbar spine mimicking a spinal meningioma. Eur Spine J. 2018;27:815–9.
9 Sacko O, Haegelen C, Mendes V, Brenner A, Sesay M, Brauge D, *et al*. Spinal meningioma surgery in elderly patients with paraplegia or severe paraparesis: a multicenter study. Neurosurgery. 2009;64:503–9; discussion 9–10.
10 Gottfried ON, Gluf W, Quinones-Hinojosa A, Kan P, Schmidt MH. Spinal meningiomas: surgical management and outcome. Neurosurg Focus. 2003;14:e2.
11 Gelabert-Gonzalez M, Garcia-Allut A, Martinez-Rumbo R. [Spinal meningiomas]. Neurocirugía. 2006;17:125–31.
12 Abul-Kasim K, Thurnher MM, McKeever P, Sundgren PC. Intradural spinal tumors: current classification and MRI features. Neuroradiology. 2008;50:301–14.
13 Rokni-Yazdi H, Sotoudeh H. Prevalence of "dural tail sign" in patients with different intracranial pathologies. Eur J Radiol. 2006;60:42–5.
14 Bansal D, Diwaker P, Gogoi P, Nazir W, Tandon A. Intraparenchymal Angiomatous meningioma: A diagnostic Dilemma. J Clin Diagn Res. 2015;9:ED07–8.
15 Setzer M, Vatter H, Marquardt G, Seifert V, Vrionis FD. Management of spinal meningiomas: surgical results and a review of the literature. Neurosurg Focus. 2007;23:E14.
16 Gezen F, Kahraman S, Canakci Z, Beduk A. Review of 36 cases of spinal cord meningioma. Spine (Phila Pa 1976). 2000;25:727–31.
17 Mawrin C, Perry A. Pathological classification and molecular genetics of meningiomas. J Neurooncol. 2010;99:379–91.
18 Lee GC, Choi SW, Kim SH, Kwon HJ. Multiple extracranial metastases of atypical meningiomas. J Korean Neurosurg Soc. 2009;45:107–11.